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Case Report

3D print model for surgical planning in a case of recurrent osteoblastic osteosarcoma of the left maxilla. A case report☆

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ABSTRACT

Osteosarcoma (OS) of the head and neck is a rare and aggressive disease characterized by the formation of osteoid by malignant osteoblasts. The mandible or maxilla are the most common sites of presentation. Radiologically, these tumors show considerable, destructive growth with periosteal reaction, which can suggest the diagnosis of OS. 3D printing, as an emerging technology, can play a role in orthopedic oncology by providing patient-specific 3D printed models to improve surgical planning and facilitate patient understanding.

We present the case of a male in his early 30s with a final histological diagnosis of recurrent osteosarcoma of the left maxilla, where a 3D printed model was helpful for the diagnostic workup, surgical planning, and the procedure.

Introduction

Osteosarcoma (OS) of the head and neck is a rare disease with aggressive behavior characterized by the formation of the osteoid by malignant osteoblasts [1]. The mandible or maxilla are the most common sites of presentation [1–4]. Radiologically, these tumors show considerable, destructive growth with periosteal reaction, which can suggest the diagnosis of OS [5]. Local recurrences predominate in head and neck osteosarcoma, with a reported incidence of 17%-70%, compared to 5%-7% in limb osteosarcoma [6]. It has been reported that 3D printing technology can play an essential role in orthopedic oncology, providing patient-specific 3D

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printed models improving surgical planning, and facilitating patient understanding [2,3].

We present the case of a male in his early 30s, with a final histological diagnosis of recurrent osteosarcoma of the left maxilla, where a 3D printed model was helpful in a multidisciplinary setting for the diagnostic workup and pretreatment surgical planning.

Case summary

This case involves a 32-year-old man who presented to the clinic with complaints of diplopia and left exophthalmos. The patient had a personal history of osteoblastic osteosarcoma of the left maxilla, which was treated with neoadjuvant chemotherapy (doxorubicin and cisplatin) with an incomplete response. The patient subsequently underwent surgery, with complete left maxillectomy and free margins confirmed by histopathology. At physical examination, all vital signs were normal. A palpable tumor in the left upper maxillary region was detected, and no palpable nodules in the neck or oral cavity alterations were identified. The laboratory work-up revealed all normal values. A contrast-enhanced head and neck computed tomography, as well as a contrast-enhanced MRI of the head, were requested to diagnose and stage the tumor.

Imaging findings

The patient's medical history includes prior surgery for osteosarcoma involving the left upper maxillary sinus. Current contrast enhanced computerized tomography (ceCT) imaging reveals recurrent growth with concerns regarding involvement of the rest of the left maxilla, the ethmoid bones, and the ipsilateral orbit, identifying a 58 mm new solid mass characterized by heterogeneous contrast-enhancement, invading the left orbital floor, also notice the absence of the maxillary sinus walls corresponding with postsurgical changes (Figs. 1A–D).

The contrast-enhanced magnetic resonance imaging (ceMRI) better depicted the extension of the solid mass lesion, sized 61 mm, and the soft tissue involvement, mainly the orbital muscles, and safely ruled out the optic nerve involvement (Fig. 2A and B).

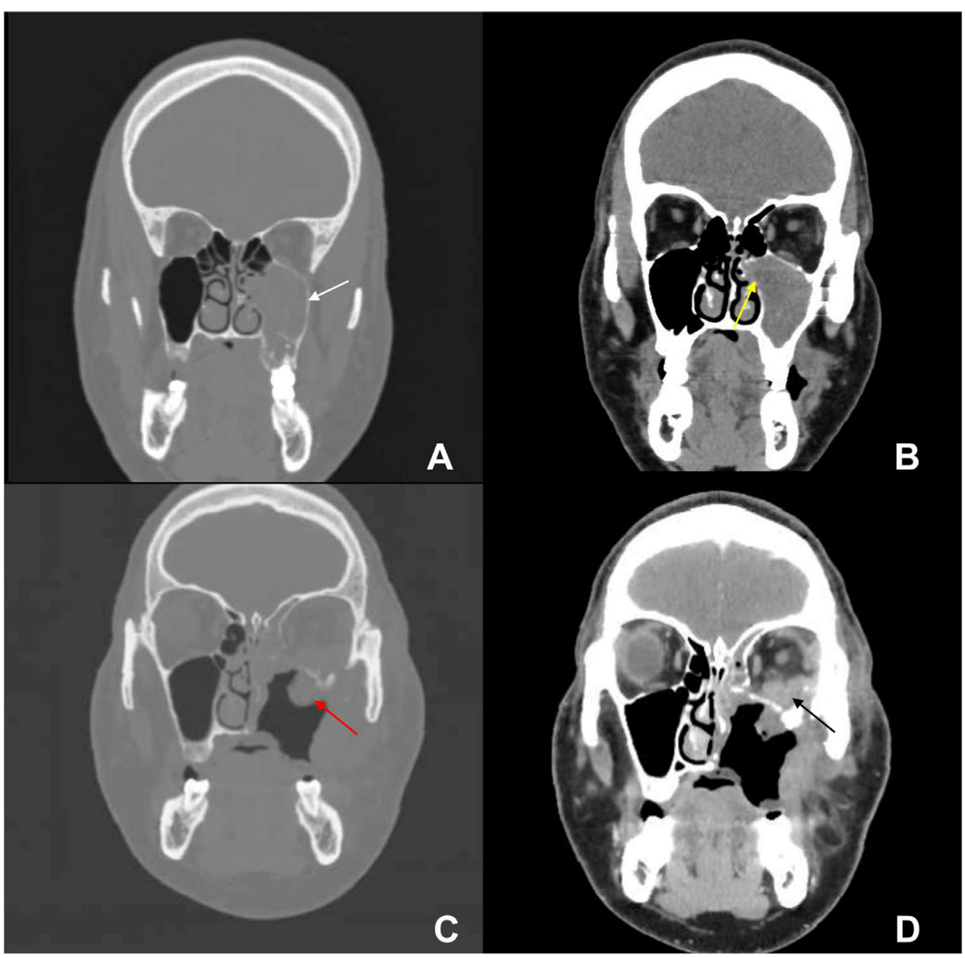

Fig. 1 – (A–D) Previous ceCT (A and B) left maxillary sinus occupied by heterogeneous soft tissue mass (white arrow), calcifications and heterogeneous enhancement, lytic changes, and invading nasal cavity (yellow arrow). Current ceCT (C and D) postsurgical changes and a recurrent soft tissue mass (red arrow) invading the left orbit cavity (black arrow).

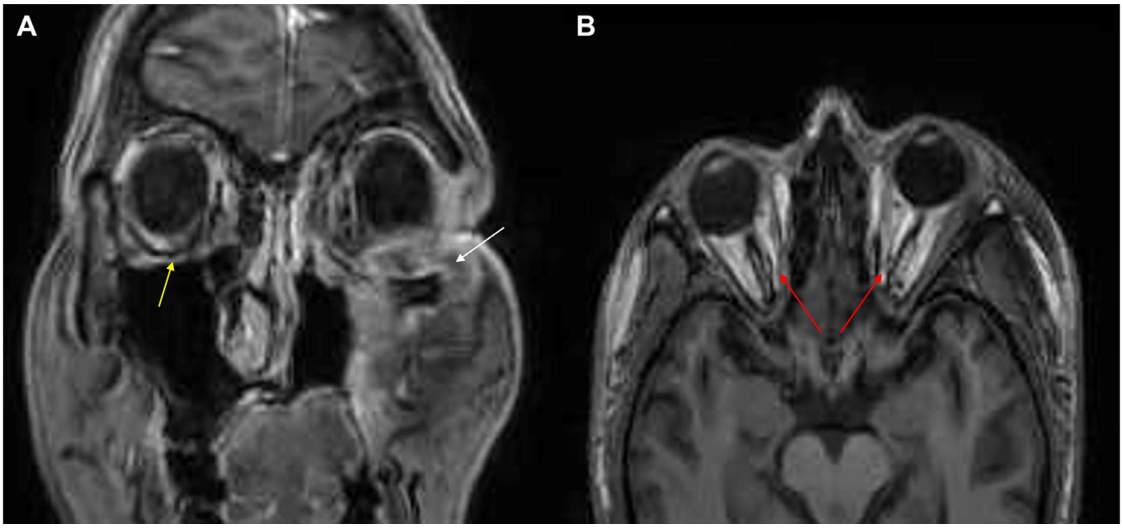

Fig. 2 – (A–B) CE-MRI Showing in the left maxillary region, (A) coronal T1 postcontrast, a soft tissue tumor extends through the orbit floor and infiltrates the inferior rectus muscle (white arrow), intact right inferior rectus muscle (yellow arrow) (B) axial 2D-FSPGR image visualizes both intact optic nerves (red arrows).

A CT-guided biopsy was performed with the diagnosis of local recurrence of osteoblastic osteosarcoma with high nuclear grade in the left maxilla.

To aid in the treatment planning process, a 3D-printed model of the solid mass lesion was created from the patient ceCT, segmented using 3D-Slicer software (Fig. 3A–D).

A fused 3D-virtual model was created, using ceCt and ceMRI images. The resulting model provided the most precise information regarding the bone structures from the ceCT and soft tissue components from the ceMRI (Figs. 4A and B).

To differentiate typical structures surrounding tumor mass lesions, the ceCT 3D model was printed in PLA material using 3 different colors. The Fused images 3D model was also printed in PLA material conformed by 5 different colors (Figs. 5A–C). This allowed for a more accurate representation of the structures and aided in the treatment planning process. The surgeon used the 3D virtual and printed models for surgical procedure planning and to explain the complexity of the disease to the patient. The physical model was also helpful in defining the best site to anchor the osteosynthesis material and fibula graft reconstruction.

The patient eventually underwent a wide local excision (Fig. 6), with a final histology diagnosis of high-grade, recurrent osteoblastic osteosarcoma. The postoperative course was favorable.

However, another recurrent lesion was found in the soft palate 2 months later, with progression through the nasopharynx and right orbit despite 1 cycle of gemcitabine (1000 mg/m^2 on days 1 and 8 from a 21-day cycle). We documented a >16% loss of heterozygosity, and the patient was enrolled in a phase 2 clinical trial for the agnostic use of Olaparib and pembrolizumab. Treatment adherence was good despite the swallowing sequelae after the wide excision. Still, local, and pulmonary progression was documented after only 2 cycles, and the patient passed away 1 month after trial exit and 4 days of palliative sedation.

Discussion

Osteosarcomas are aggressive bone tumors that primarily affect the long bones but can also occur in the head and neck region, although less frequently. Head and neck osteosarcomas pose unique challenges in diagnosis and management due to the complex anatomy and critical structures involved. Jaw osteosarcomas, specifically in the mandible, are even rarer, accounting for less than 1% of head and neck cases. The incidence of osteosarcoma is 2-3 million per year, with a higher occurrence in adolescence (8-11 million per year in the 15-19 age group). Typical symptoms include local pain, localized swelling, and limited joint movement [1–4,6–9].

Radiologically, these tumors exhibit substantial and aggressive growth patterns, characterized by extensive destruction of surrounding tissues and prominent periosteal reactions. These findings strongly support the diagnosis of osteosarcoma (OS), a malignant bone tumor known for its invasive nature and ability to cause significant osseous destruction [6].

In plain radiograph, the typical appearance of high-grade osteosarcoma includes aggressive destruction of medullary and cortical bone, a wide transition zone, a permeative appearance, aggressive periosteal reaction, periosteal reaction as onion skin and sunburst pattern, sometimes with radiating spicules extending from the tumor. The presence of the Codman triangle is another characteristic feature [10,11]. CT is primarily used for supporting biopsy procedures and staging of tumors. For predominantly lytic lesions, small amounts of mineralized material bring additional information not viewed in another imaging modality [11,12]. MRI is an essential tool to determine accurate local staging and evaluation of intraosseous tumor extension and soft-tissue involvement. Very useful for evaluating the growth plate and if the epiphysis is compromised [12].

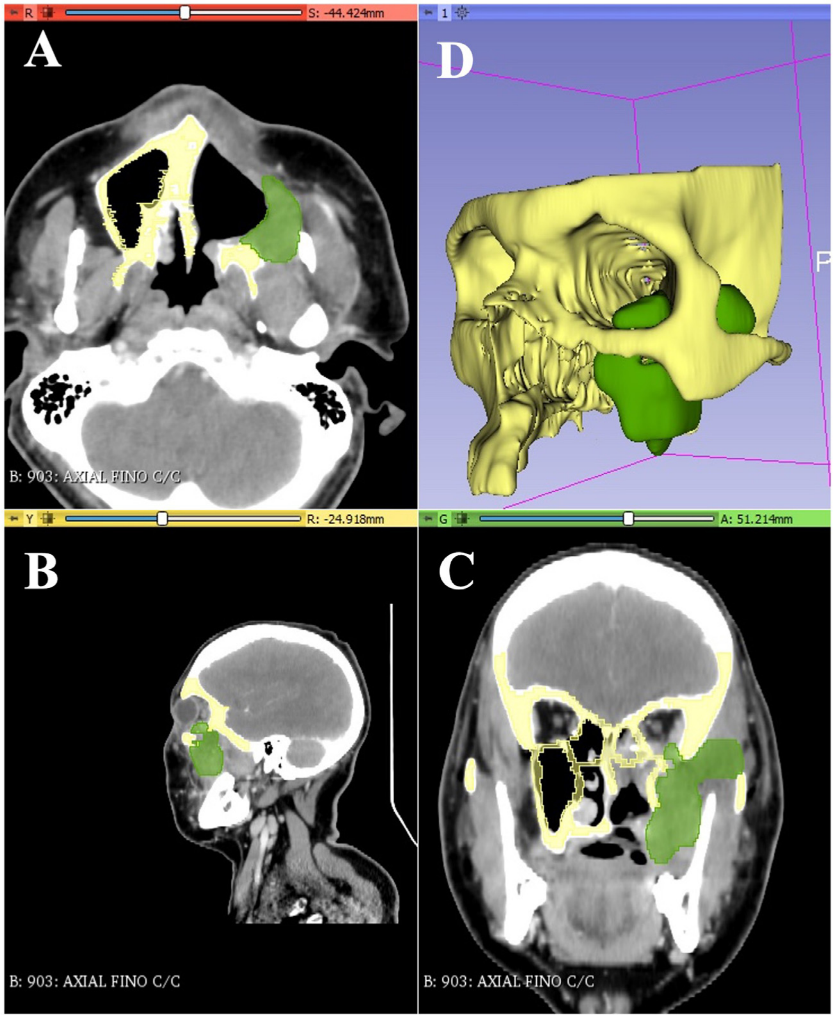

Fig. 3 – (A–D). CT images, (A) Axial plane reconstruction, (B) sagittal plane, (C) Coronal plane, processed using 3D-slicer program to create (D) 3D-model to be printed, manually segmented by Radiologist for a better detail visualization, lesion tumor in green, and bone structures in yellow.

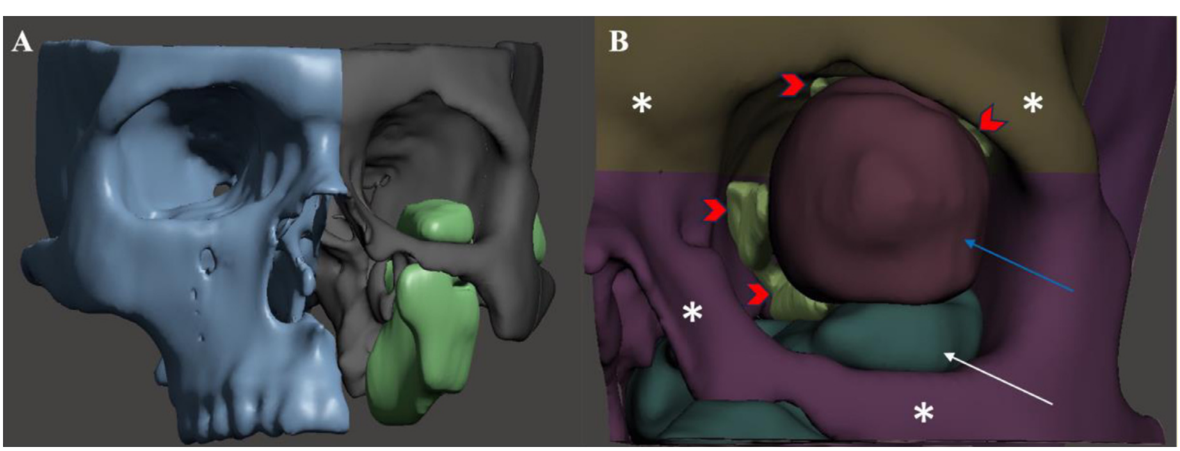

Fig. 4 – (A–B) Fused ceCT and MRI images provide accurate evaluation and comparison. (A) healthy right side in blue and affected (left) bone structures in gray and mass lesion in green, (B) affected left side, showing extraocular muscles (Red arrowheads), bone extructures (white asterisk), mass lesion (withe arrow), eyeball (blue arrow).

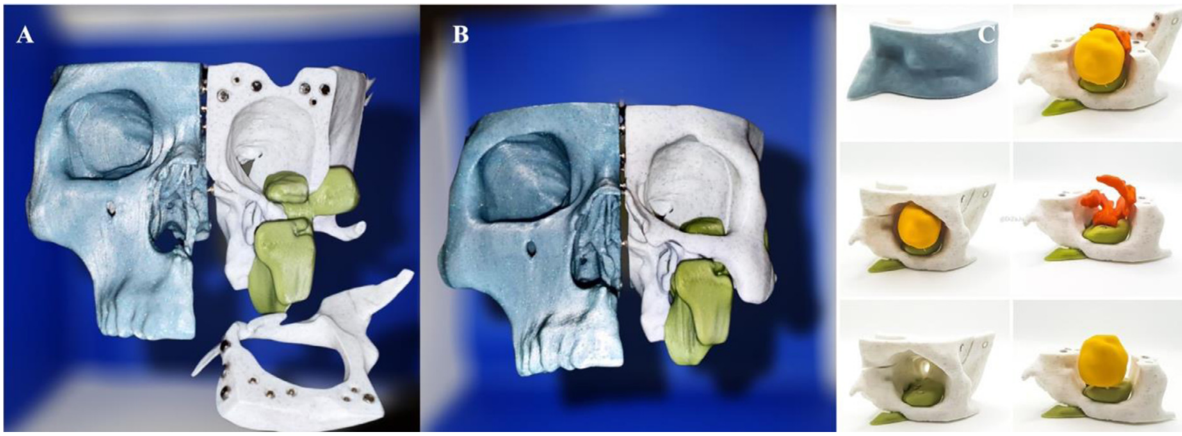

Fig. 5 – (A–C). (A and B) 3D-model healthy right facial bones blue, left soft tissue mass lesion green, left bone affected structures white, (C) fusion 3D-model, left skin blue, bone structures white, eyeball/optic nerve yellow, orbital muscles red, solid mass green.

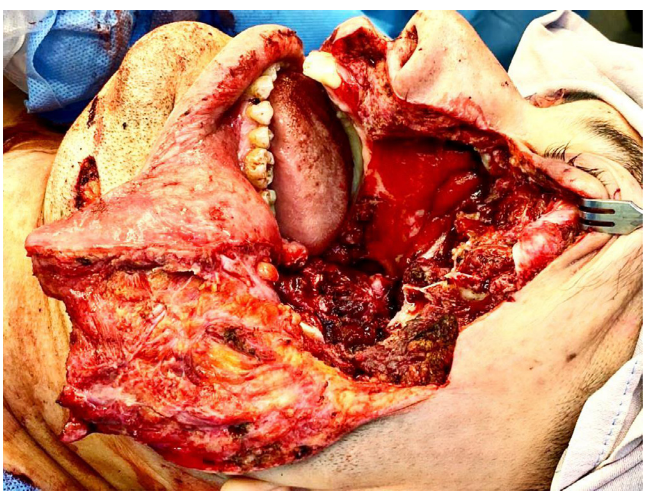

Fig. 6 – The intraoperative image demonstrates the surgical site after the removal of the tumor lesion.

In our discussed case, a patient presented with recurrent osteosarcoma in the left maxillary sinus, with concerns regarding potential infiltration into ophthalmic structures, particularly the optic nerve. However, a contrast-enhanced MRI and the utilization of 3D imaging techniques ruled out such involvement and provided a better understanding of the disease's limits and extent, indicating the affection of only the extraocular muscles. This information proved valuable for surgical planning and ensuring precise resection boundaries.

Fused 3D imaging, combining CT and MRI, is increasingly used in orthopedic oncology. It enables the creation of patient-specific 3D printed models, customized surgical guides, and implants, enhancing surgical planning. Collaboration among radiologists, orthopedic surgeons, and engineers is crucial for successful 3D printing. Studies, such as Punyaratabandhu et al., have demonstrated the benefits of 3D printing in tumor cases, aiding in personalized pre-operative planning, physical simulations, and guiding complex surgeries. These advantages include reduced blood loss, shorter operative time, and optimal selection of surgical approaches and implants [3,5,13–16].

Treatment for osteosarcoma includes surgery with perioperative chemotherapy. Advances in systemic therapy have significantly improved overall survival rates, with limb osteosarcoma achieving 5-year survival rates of 60%-70%. Surgery alone or in combination with radiotherapy can yield survival rates of up to 85%. Notably, complete resection remains a key factor in the successful management of low-grade osteosarcomas [1,8,17]. The free vascularized fibula flap constitutes the most popular surgical technique for mandibular reconstruction in young patients, first described in 1975 and first reported in the literature in 1985. This procedure necessitates a high level of precision and comprehension of the disease in order to accurately identify the osteotomy site and secure the osteosynthesis material [2,3,13].

Head and neck osteosarcomas exhibit unique recurrence patterns, with a higher incidence of local recurrence compared to other types of osteosarcoma. The challenges associated with achieving tumor-free margins through surgical intervention contribute to the increased rates of local recurrence in this region [2].

Conclusion

In conclusion, head, and neck osteosarcomas, although rare, pose unique diagnostic and management challenges. Radiological imaging, including fused 3D imaging techniques, plays a crucial role in assessing disease extent and guiding surgical planning. The integration of 3D printing technology provides personalized benefits and aids in achieving improved patient outcomes. Collaborative efforts among multidisciplinary teams are essential.

Declaration of generative AI and AI-assisted technologies in the writing process

During the preparation of this work, the author(s) utilized Chat-GPT to review the language and enhance readability. Following the use of this tool/service, the author(s) thoroughly reviewed and edited the content as required and assume(s) full responsibility for the publication's content.

Patient consent

We confirm that written, informed consent for the publication of this patient's case was obtained prior to dissemination of any information.

The patient, was fully informed of the nature and purpose of the case study and the potential use of their medical information, including their personal and medical history, for educational and research purposes.

The patient was informed that their name and any other personal identifiers will be removed or obscured in the publication to maintain confidentiality.

The patient could ask any questions and fully understands their rights.

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